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Case Report

Rare and Unusual Localization of Adenoid Cystic Carcinoma: A Case Report

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Abstract

Introduction:

Adenoid cystic carcinoma (ACC) of the external auditory canal (EAC) is an extremely rare tumor.

Case Report:

This is about a 62-year-old female Moroccan patient with no particular medical history who consulted for a mass in the right external auditory canal revolving for 1 year. The biopsy showed an ACC of the right external auditory canal.

Discussion and Conclusion:

The external auditory localization of ACC is rare and unusual, making diagnosis and treatment difficult given the low incidence of the disease. A pathological examination must be performed to establish a positive diagnosis and assess the margins. For patients with uncertain negative margins, postoperative radiotherapy and chemotherapy may be considered to reduce recurrence and improve survival.

Keywords: adenoid cystic carcinoma ; external auditory canal ; anatomical pathology

Introduction

Adenoid cystic carcinoma (ACC) of the external auditory canal (EAC) is an extremely rare tumor (1). First described as "cylindroma" by Billroth (2), ACC is generally classified with salivary gland tumors, although it can appear in any site where mucous glands exist. Half of these tumors occur in glandular areas other than the major salivary glands, primarily in the hard palate, but also in the tongue and other areas with minor salivary glands [3,4]. The external auditory canal is an unusual location [5,6]. We report a case of ACC of the external auditory canal.

Medical Observation :

This is about 62-year-old female Moroccan patient with no particular medical history who consulted for a mass in the right external auditory canal evolving for 1 year with intermittent otalgia and occasionally blood-stained discharge on friction without other associated auditory or other signs. Clinical examination showed a mass in the right external auditory canal without inflammatory signs and non-painful on palpation. The tympanic membrane was obscured by the mass. The rest of the ENT, lymph node, and general examination was unremarkable. A CT scan of the temporal bones revealed a solid, round mass of medium density enhanced by contrast. The neoplasm measured 1.5 cm and was located in the upper-anterior part of the right external auditory canal. A biopsy was performed, showing a biphasic tumor proliferation of epithelial and myoepithelial cells suggestive of ACC (figures 1,2). A multidisciplinary consultation meeting was held, and the therapeutic decision was to perform a modified right mastoidectomy.

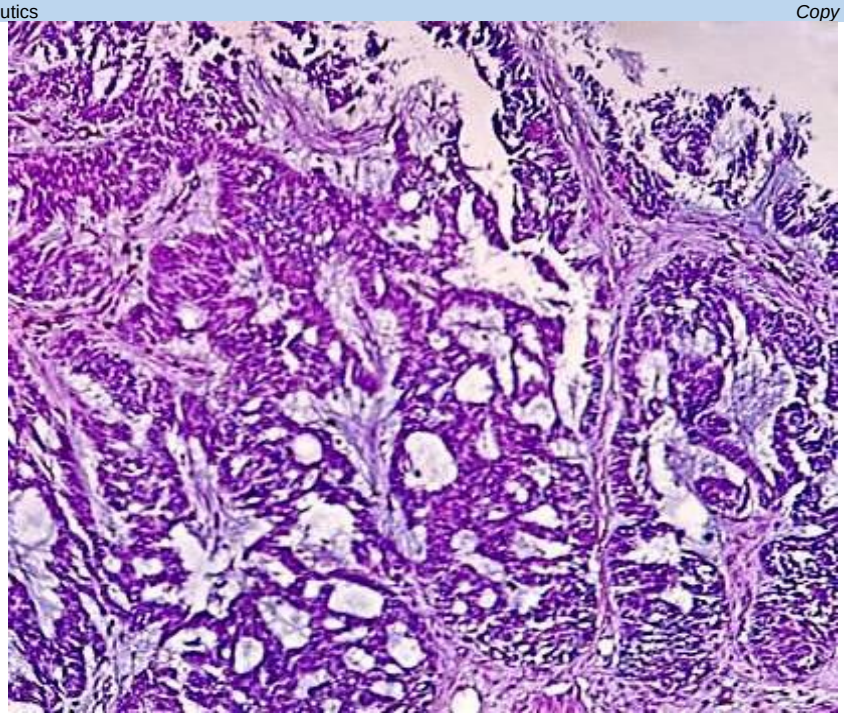

Figure 1: Biphasic tumor, composed of ductal and myoepithelial cells

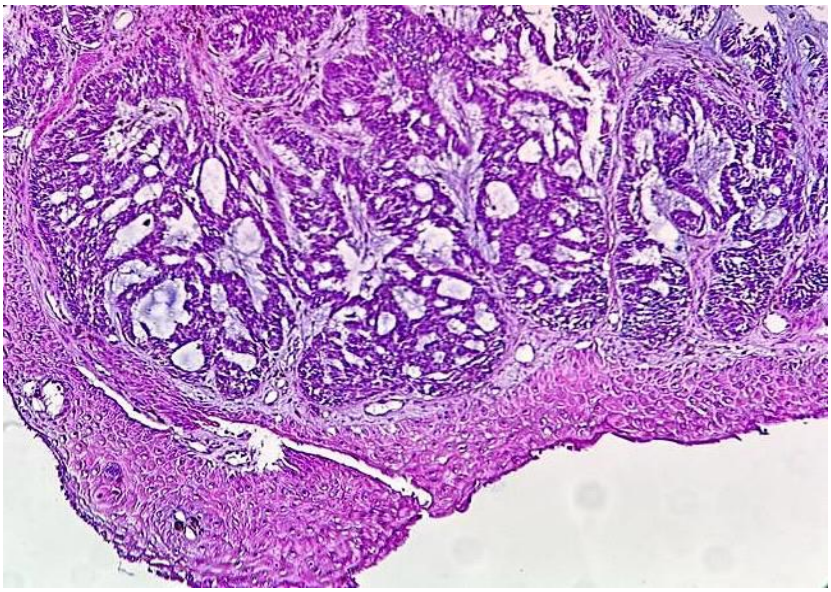

Figure 2: Cribriform pattern composed of predominantly myoepithelial cells with myxoid or hyalinized globules

Discussion

Malignant tumors of the external auditory canal are rare, most being squamous cell carcinomas. Adenocarcinomas developing in the external auditory canal are extremely rare. Although ACC is a rare tumor in the external auditory canal, it is relatively common in the salivary glands of the head and neck. Clinical symptoms of ACC of the external auditory canal can be nonspecific, usually presenting with hearing loss, masses in the external auditory canal, otorrhea, and otalgia. Early intermittent otalgia is the most common symptom in patients with ACC of the external auditory canal, but infection and otorrhea are not uncommon in some elderly patients. The main differential diagnosis at this stage is chronic otitis media. ACC has three main histological patterns: tubular, cribriform, and solid. In salivary glands, the prognosis of ACC is correlated with the predominant histological profile. Tubular ACC has the best prognosis, while solid ACC has a worse prognosis (10). Four series are reported in the literature (6) (7) (8) (9), and the others are case reports ranging from one to three cases. The surgical principle for the tumor is the same as for most malignant tumors, which is complete curative resection. Surgical methods include local resection of the EAC, en bloc resection of the EAC, subtotal temporal bone resection, and temporal bone resection. A positive margin is a vital factor leading to a poor prognosis (11). Additionally, treatment of the parotid gland is controversial. Some authors consider that superficial parotidectomy is suitable for all patients with ACC of the EAC, even in early stages (12). Radiotherapy can destroy subclinical tumor foci and increase surgical efficacy. Radiotherapy can be particularly effective for advanced tumors

when safe margins are difficult to achieve (13). Postoperative radiotherapy is necessary for patients with advanced clinical disease, while patients with negative margins after the first surgery and low clinical stage do not need radiotherapy (14). The survival rate was significantly higher for patients who received surgery and radiochemotherapy than for patients who did not receive all three treatment modalities in some studies (6).

Conclusion

In cases of recurrent chronic otitis media or chronic and recurrent auditory symptoms, tumor pathology of the external auditory canal should be considered. Imaging and biopsy should be performed quickly to establish the diagnosis. It is recommended to expand the tumor for the first operation and perform a superficial parotidectomy. A pathological examination must be performed to establish a positive diagnosis and assess the margins. For patients with uncertain negative margins, postoperative radiotherapy and chemotherapy may be considered to reduce recurrence and improve survival.

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Availability of data and materials

Data available

Declarations

Ethics approval and consent to participate

The patient authorizes the publication of this article.

Consent for publication

Written informed consent was obtained from the patient for publication of this case report and any accompanying images.

Competing interests

The authors declare no competing interests.

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